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Case Report

Intraparenchymal Pericatheter Cyst as a Complication of a Ventriculo-Peritoneal Shunt in a Premature Infant

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A ventriculo-peritoneal shunt is a standard surgical management for hydrocephalus, but complications may impede the management of this disease. Obstruction of the catheter is one of the most common complications and manifests clinically in various ways. Intraparenchymal cyst development after shunt malfunction has been reported by several authors, but the underlying mechanism and optimal treatment methods are debatable. The authors report a case of intraparenchymal cyst formation around a proximal catheter in a premature infant after a ventriculo-peritoneal shunt and discuss its pathogenesis and management.

Key Words : Intraparenchymal cyst · Ventriculo-peritoneal shunt · Complication · Premature infant.

INTRODUCTION

Since the introduction of cerebrospinal fluid (CSF) shunting procedures, they have been one of the main methods of managing hydrocephalus. However, complications of shunt surgery may impede the management of hydrocephalus, and have been reported to affect about 20 to 30 percent of cases during long term follow up[11,12]. Infection and obstruction are the most common complications, though improvements in shunt valve design have diminished complication rates caused by nonphysiologic hydrodynamics of the shunt system. The spread of CSF into brain parenchyma is a rare complication of a ventriculo-peritoneal shunt[2,5,10,14,18], and can take the form of CSF edema or a reversible porencephalic cyst[13].

Pericatheter cysts have been described in relation to a blocked shunt in children and adults, but it has not been previously reported in a premature infant. The authors present a case of an intraparenchymal CSF cyst in a premature infant who happens to be the youngest patient ever reported with this condition.

CASE REPORT

An one-day-old male infant was admitted to our neonatal intensive care unit due to weak crying and whole body cyanosis. He was born at gestation week 28 to a 30-year-old multipara at our hospital by Cesarean section delivery due to premature rupture of the maternal membrane. His birth weight was 1,450 g and his Apgar scores were 6 and 9 at 1 and 5 minutes, respectively. After birth, he was provided positive pressure ventilation and intubated. He also suffered respiratory distress and was treated with surfactant and by mechanical ventilation. On the 24th day after birth, he was presented with fontanel bulging with slight activity reduction. Moro reflex was partial but symmetric, and his pupils were of equal size and reactive to light bilaterally. Ultrasonography and MRI of the brain revealed severe hydrocephalus (Fig. 1). External ventricular drainage was performed, and 8 days later at a body weight of 1,600 mg, a ventriculo-peritoneal shunt was inserted using contoured-shape ultrasmall PS Medical vlave. Two months after shunt surgery, follow-up CT and MRI showed pericatheter ventricular cyst formation (Fig. 2). Burr hole drainage of the pericatheter cyst was performed, and 80 cc of a yellowish translucent fluid was aspirated. Fluid analysis revealed, a cell count of 6/uL (polymorphous nuclear cells 17%), glucose 36 mg/dL, protein 2633 mg/dL, and gram stain negativity, and culture showed no growth of bacteria and fungus. The drain catheter was removed on the 9th day after insertion and a follow up CT scan revealed near total cyst disappearance. Two months later, head enlargement and a tense fontanel were noted, and follow up MRI revealed intraparenchymal cyst regrowth with normally sized ventricle. Because reinsertion of the new proximal catheter into the ventricle was considered difficult without direct visualization, craniotomy and shunt revision was planned.

Craniotomy and opening of the cyst revealed a pseudocyst

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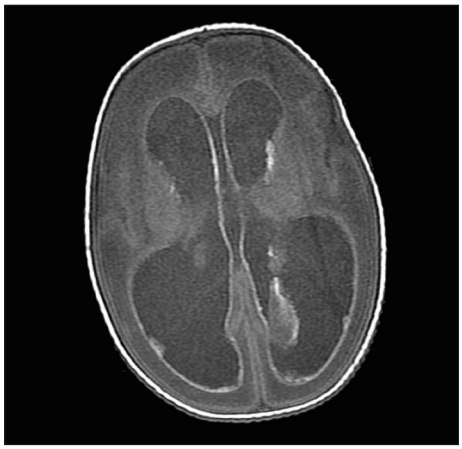

Fig. 1. Initial axial T1-weighted MR images showing hydrocephalus.

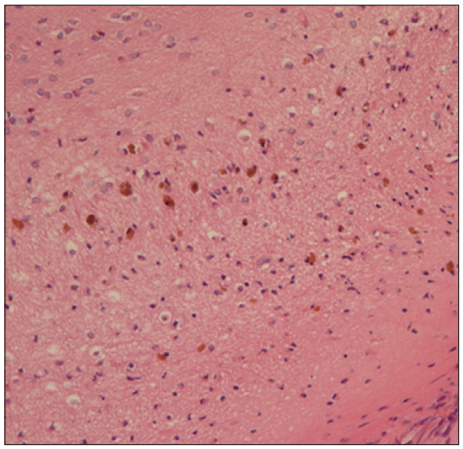

Fig. 3. Pathological examination of the cystic wall revealing a pseudocyst and reactive gliosis.

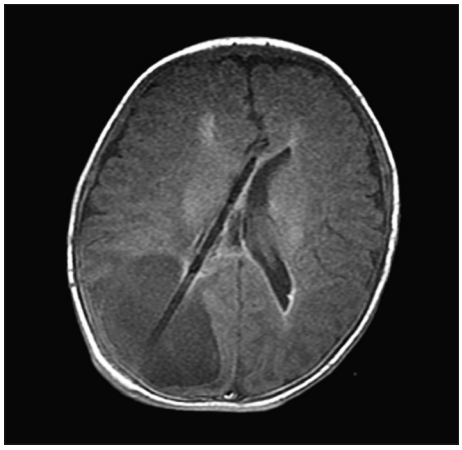

Fig. 2. Axial T1-weighted MR images taken 2 months after initial shunt placement showing the pericatheter intraparenchymal cyst.

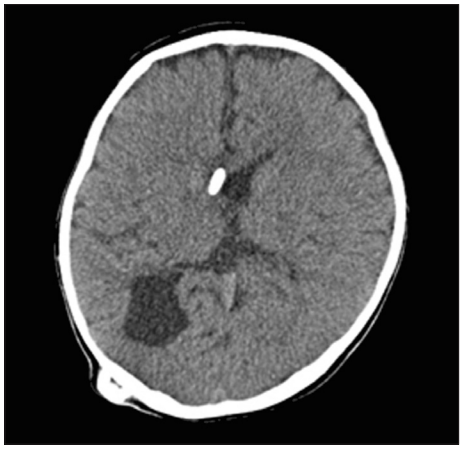

Fig. 4. Follow up CT scan taken 16 months after shunt revision showing a remnant cyst around the catheter and no pressure effect.

with a gliotic wall and the proximal shunt catheter passing through the cyst. The deep portion of the cyst was separated from the ventricle by thin partially transparent ependymal and parenchymal tissues. There was a small amount of pericatheter CSF leakage. Biopsy of the cyst wall showed reactive gliosis (Fig. 3), and an evaluation of the inside of the shunt catheter showed partial proximal obstruction due to brain tissue-like material. The distal catheter was found to be patent. The shunt was replaced with a Strata valve (small size, level 0.5, Medtronic, USA). Follow up CT scan taken 16 months after shunt revision revealed a small residual cyst (Fig. 4).

DISCUSSION

Germinal matrix-intraventricular hemorrhage is the most common type of neonatal intracranial hemorrhage and is a characteristic complication of premature delivery[7,19]. About 20-50% of cases require a ventriculo-peritoneal shunt due to hydrocephalus[7,8,20]. In many such cases, posthemorrhagic hydrocephalus resolves spontaneously without intervention. However, if serial cranial ultrasonographic examinations show persistent posthemorrhagic ventricular dilatation, intervention is usually required. Ventriculo-peritoneal shunt is regarded as the standard therapy for the management of hydrocephalus, even though some have reported that endoscopic third ventriculostomy is useful alternative method.

Shunt placement in small premature infants carries the risk of considerable morbidity, secondary to various shunt-related complications, such as, ulceration of scalp, ventriculitis, sepsis, and necrotizing enterocolitis, and the need for frequent revision[3,9,21]. Various temporary procedures have been used to delay ventriculo-peritoneal shunt placement, for example, subcutaneous reservoir embedment surgery, a ventriculo-subgaleal shunt, external ventricular drainage, and repeated lumber puncture; however, success rates vary[4,21,22].

The occurrence of a pericatheter CSF cyst has been reported by several authors. Vajramani and Fugleholm[18] reported an intraparenchymal CSF cyst in a patent shunt patient after radiation therapy due to a brain tumor; it was hypothesized that the radiation therapy contributed to cyst development. In this case, no obstruction of the catheter system was observed, but an observed improvement after catheter replacement suggested par-

Table 1. Summary of reported intraparenchymal cysts after ventriculo-peritoneal shunt placement

Reference number	Number of cases	Age	Interval from shunt and cyst formation	Primary disease	Management	Outcome
5	2	10 Y	2 Y	Myelomeningocele	Shunt revision	Improved
		10 Y		Posttraumatic	Shunt revision	Improved
15	2	4 Y	3 Y	Myelomeningocele	None	Improved
		5 M	1 M	Intraventricular cyst	None	Improved
16	4	5M	2 M	Myelomeningocele	Shunt revision	Improved
		2 Y	2 Y	Encephalocele	Shunt revision	Improved
		3 Y	2 Y	Meningitis	Shunt revision	Improved
		10 Y	7 M	Acute lymphocytic leukemia*	VP shunt	Improved
18	1	51 Y	4 Y	Meningioma	Shunt revision	Improved

*Ommaya reservoir insertion. M : months, Y : years, VP : ventriculo-peritoneal

tial catheter obstruction.

We postulate that CSF leaked through the pericatheter space due to increased intracranial pressure after partial obstruction of the shunt catheter, and the one way check valve mechanism of the ventricular wall causes cyst enlargement. In our case, the obstruction was found in the proximal region of the ventricular catheter, and we believe that the relatively soft brain parenchymal consistency of prematurity contributed to cyst development. However, there is controversy about the etiological hypothesis. Sinha et al.[15] reported two intraparenchymal pericatheter cyst cases following ventriculo-peritoneal shunt placement, but in these cases cyst-related symptoms were absent and they were followed using serial CT scans, which revealed progressive decreases in cyst sizes. However, the wait-and-see management approach requires that special attention be paid to the development of symptoms, which is not easy in young patients, and that serial CT or MRI be performed. Unfortunately, the repeated CT scans are not considered a safe option in young children due to the risk of delayed cancer development in the irradiated area[1,6,17].

A literature search of the PubMed database using the keywords, ventriculoperitoneal, cyst, and porencephaly, revealed 4 articles on 9 patients (Table 1)[5,15,16,18]. Eight of nine patients were between 5 months and 10 years of age; the other exceptional patient was 51 years old. Durations from shunt surgery to cyst identification ranged from 1 month to 4 years. Most had a shunt obstruction and shunt revision was the most relevant management modality.

Iqbal et al.[5] described that a greater pressure gradient between ventricle and pericatheter space and degree of gliosis along the catheter increased the risk of cyst development. However, our patient was a premature infant, and thus, the brain was soft and the cyst was able to expand easily around the catheter.

Sakamoto et al.[13] reported four cases of pericatheter edema after placing a ventriculo-peritoneal shunt, though all four improved after shunt revision. The pathogenesis of pericatheter edema is regarded to be similar to that of cyst formation. Sakamoto et al.[13] reported cases with the inflow holes of the catheter tip partially lying in the intraparenchyma outside the lateral ventricle producing pericatheter edema. Special care is required when the proximal catheter is inserted through Kocher's point[13]. Chiba et al.[2] also reported CSF edema as a shunt complication and all of their patients had a shunt obstruction. The determinator of cyst or edema formation is unclear, although the speed of CSF flow and brain consistency are likely to be related with the differential development of edema or cyst as a result of CSF flow into the brain. Our hypothesis is that the condition of the ventricular wall probably influences the course. Tight adhesion of the ventricular wall to the catheter is likely to provoke the development of edema, whereas a small aperture around the catheter would favor cyst formation. In our case, the relatively soft ventricular wall and brain consistency in a premature infant probably led to cyst formation.

CONCLUSION

Authors report a rare case of an intraparenchymal pericatheter cyst as a complication of ventriculo-peritoneal shunt in a premature infant, which is the youngest age ever reported. Partial obstruction of the proximal catheter produced the parenchymal cyst formation. Shunt revision provided the improvement of this complication.

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